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Case Report

Uterine Polypoid Hemangioendothelioma: Conservative Treatment

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Background and Objective: **A polypoid uterine hemangioendothelioma was treated by conservative means.**
Study Design/Materials and Methods: **The diagnosis of hemangioendothelioma was confirmed by histopathology and angiography. The patient desired to retain her fertility; therefore, the hemangioendothelioma was treated by hysteroscopically guided Nd:YAG laser ablation.**
Results: **Five years of follow-up by hysteroscopy and dilation and curettage reveal no evidence of recurrence. The patient continues to have regular menses.**
Conclusion: **Conservative management of a uterine hemangioendothelioma by Nd:YAG laser ablation was successful over 5 years of follow-up.**
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Key words: hemangioendothelioma, vascular myxoid leiomyoma, angiosarcoma

INTRODUCTION

We report the conservative management of a uterine hemangioendothelioma using the Nd:YAG laser.

CASE REPORT

A 20-year-old nulliparous woman presented in 1988 with a 1 1/2-year history of a continuous serosanguinous vaginal discharge. Prior to her visit, history included irregular menses present for 2 years, unsuccessful treatment with birth control pills for 4 months in an attempt to regulate her periods, and a positive chlamydia fluorescence antibody screen for which she received treatment with tetracycline.

Her physical examination was remarkable only for a serosanguinous vaginal discharge of ~8 ml per 24-hour period (collected from multiple aspirations from a cervical cap). Cytology on the fluid was negative. Ultrasound examination revealed a 1.5 × 2 cm polypoid soft tissue mass in the posterior uterine cavity with the same echogenicity as the endometrial lining. A D&C identified a fusiform 1.5 × 4 cm specimen. A preliminary histologic diagnosis of a vascular neoplasm was made. Because of the pathologic diagnosis, hysterosalpingography and pelvic angiography were performed, which showed a vascular flush consistent with a vascular malformation in the uterus (Fig. 1a–d). Laparoscopy documented the absence of additional pelvic vascular malformations, and hysteroscopy revealed a pedunculated mucoid-apearing vascular lesion of the right lateral uterine wall. Because the vessels feeding the mass did not appear to exceed the diameter of the Nd:YAG probe, ablation of the feeding vasculature was performed until the stalk was completely divided from the uterine wall. A touch technique was used in the area of the pedicle. A power setting of 60W continuous mode was used throughout.

RESULTS

The hysteroscopically removed specimen consisted of multiple irregular fragments of pale-tan rubbery tissue measuring 2.5 × 1.5 cm in aggregate. Microscopic examination revealed numerous small and intermediate-size blood vessels irregularly distributed in a myxoid stroma (Fig. 2). In addition to endothelial cells lining capillaries, many round or fusiform cells with large nuclei and well-defined cytoplasmic borders were distributed in the intervening myxoid stroma, not associated with blood vessels (Fig. 3). Both endothelial cells lining capillaries and intervening

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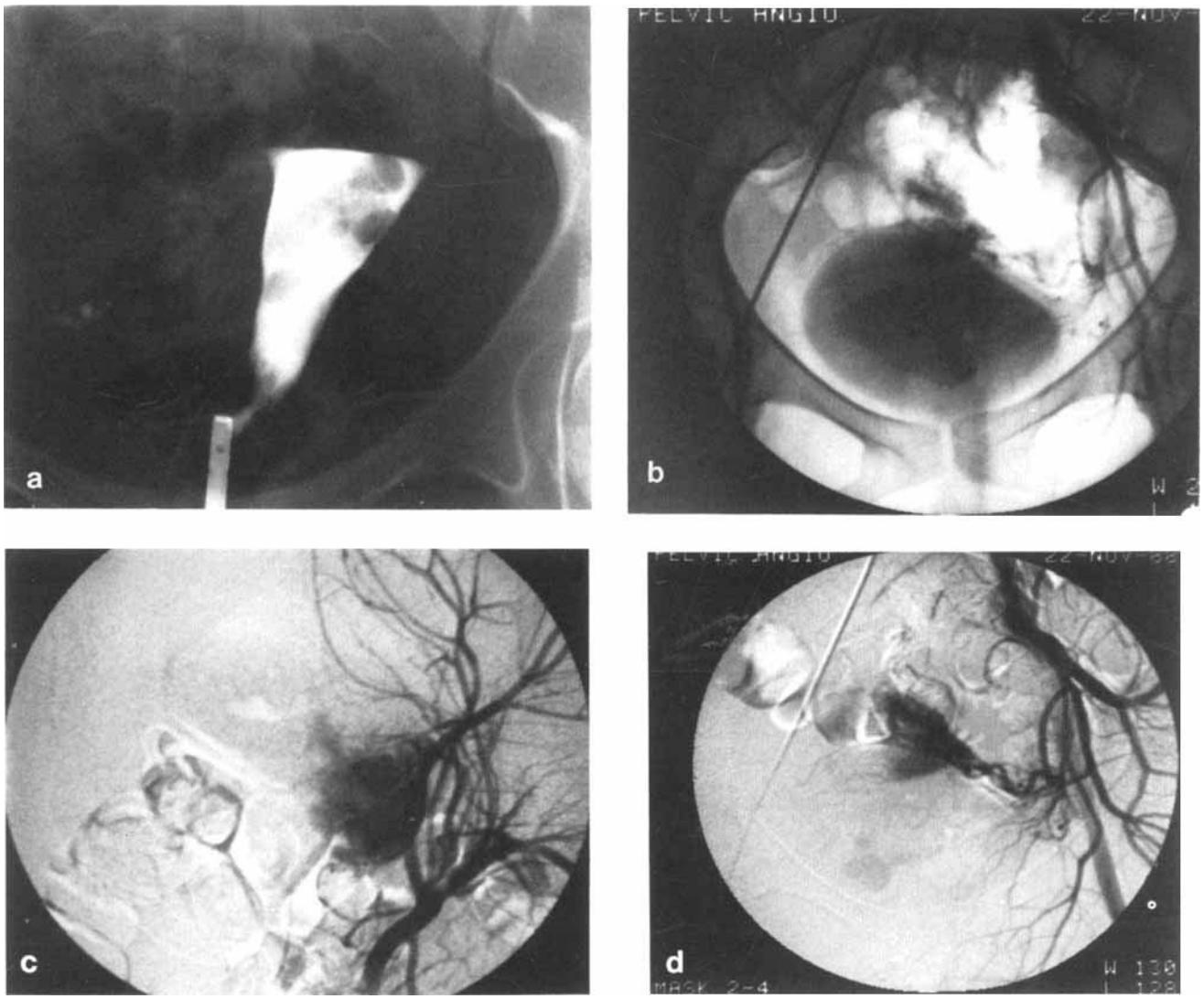

Fig. 1. (**a**) Hysterosalpingogram demonstrating a large intrauterine filling defect. (**b**) Nonsubtracted angiography demonstrating the pelvic bony architecture. (**c**) Subtracted angiography demonstrating early blushing of the hemagioendothelioma after injection of the uterine artery. (**d**) Subtracted angiography demonstrating late blushing of the hemangioendothelioma after injection of the uterine artery.

cells demonstrated mild nuclear pleomorphism. Mitotic figures were rare. The surface of the polypoid mass was lined by a single layer of endometrial epithelium that was focally denuded (Fig. 2). The histologic differential diagnosis included a hemangioendothelioma and a vascular myxoid leiomyoma.

Immunohistochemical staining revealed positive cytoplasmic reaction for factor VIII-related antigen in intervening stromal cells and in endothelial cells that lined blood vessels (Fig. 4). With the exception of the muscular wall of large blood vessels, the tumor was negative for desmin and smooth muscle myosin.

Follow-up over 5 years with hysteroscopy and biopsy has been unremarkable with no evidence of recurrence.

DISCUSSION

Vascular neoplasms of uterine corpus are uncommon [1]. Examples of both benign and malignant varieties of these tumors have been reported, but similar to vascular neoplasms of other body sites, the terminologies used are confusing. Benign vascular tumors include hemangiomas and lymphangiomas [2]. Malignant tumors include angiosarcoma, also referred to as malignant

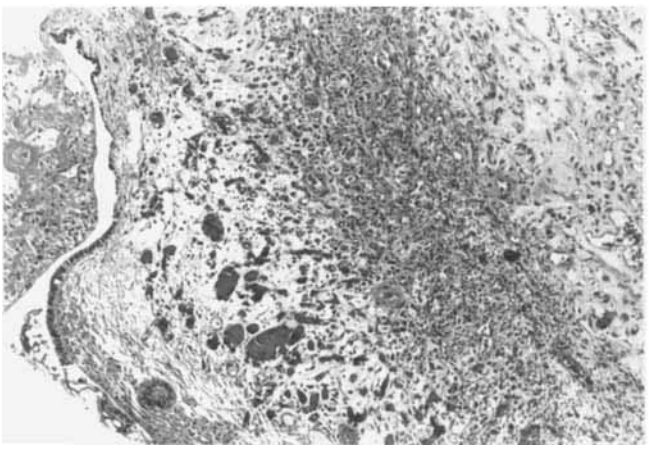

Fig. 2. Polypoid hemangioendothelioma. A single layer of endometrial epithelium is seen on the left. Many small blood vessels and capillaries are irregularly distributed in a myxoid stroma. (H&E, ×25).

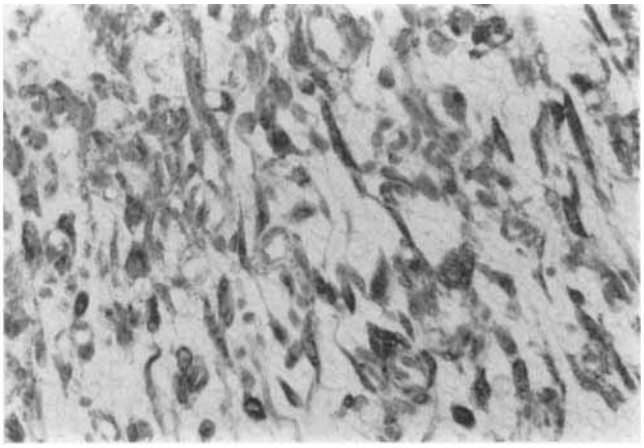

Fig. 4. Immunoperoxidase stain for factor VIII-related antigen. Both intervening cells and those lining capillaries demonstrate a granular cytoplasmic staining. (Biotin-avidin-peroxidase; light hematoxylin counterstain, ×100).

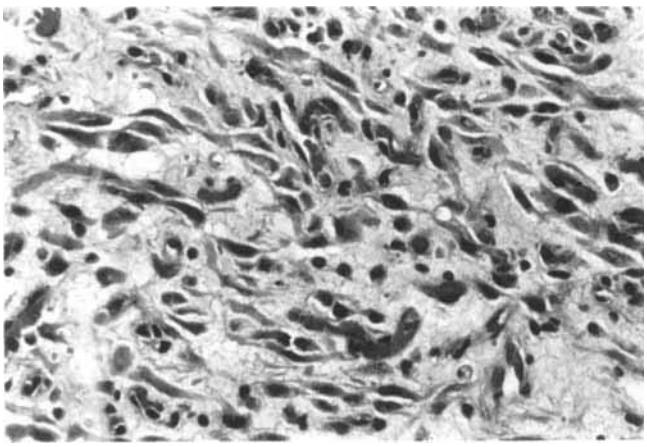

Fig. 3. Hemangioendothelioma. In addition to the endothelial cells lining capillaries, there are many isolated round or fusiform cells in the intervening stroma. (H&E, ×100).

hemangioendothelioma in older literature [3]. Between the clearly benign and frankly malignant tumors of endothelial origin, there is a heterogenous group of vascular neoplasms with a wide range of morphologies and biologic behavior [4]. These neoplasms have been referred to as hemangioendotheliomas, histiocytoid hemangiomas, and epithelioid hemangioendotheliomas among other terminologies [4]. They can potentially occur in any organ and body site, and in most cases they behave as borderline or low-grade malignant endothelial neoplasms. We believe that the current case is an example of such a neoplasm occurring in the endometrium.

The differential diagnosis of the present tumor based on morphology alone includes myxoid vascular leiomyoma and inflammatory pseudotumor of the uterus [5], both of which are rich in blood vessels. The intervening cells of vascular leiomyomas are smooth muscle cells, whereas those of inflammatory pseudotumors are myofibroblasts. Positivity of the intervening cells for factor VIII-related antigen in this case proved their endothelial nature and confirmed the diagnosis of hemangioendothelioma [6]. By contrast, the fact that these cells were negative for desmin and smooth muscle myosin excluded their smooth muscle and myofibroblastic differentiation.

Because of the rarity of this neoplasm in the uterus, its uncertain biologic behavior, and the patient's desire to retain her fertility, we chose to treat it conservatively. Follow-up over the last 5 years (May 1994) by hysteroscopy and D&C have revealed no evidence of recurrence.

In summary, this case presented with a significant serosanguinous discharge attributed to the vascular tumor. Conservative management was undertaken with the Nd:YAG laser, which has been successful for 5 years thus far with regular periods. This patient will continue to be followed by hysteroscopy and biopsy.

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